This paper reports a ten-year follow-up of 1,012 children previously studied from early in pregnancy through a two-year assessment of physical and mental handicaps of congenital origin. The findings are reported and their significance assessed.

CONGENITAL AND ACQUIRED HANDICAPS OF TEN-YEAR-OLDS—REPORT OF A FOLLOW-UP STUDY, KAUAI, HAWAII

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SEVERAL years ago we described the defects of prenatal and natal origin of two-year-old children born to mothers followed from early pregnancy.[1] Because we were dealing with an entire community's children—the island of Kauai, Hawaii—it was possible to report incidence rates of birth defects for this population of some 30,000 and estimate the community impact of such defects. With a 24 per cent probability of fetal mortality, we estimated that for each 1,000 live births there were 1,311 pregnancies which had advanced to four weeks' gestation; 286 ended in fetal death before 20 weeks' gestation and 25 more between 20 weeks and term.[2] These 1,311 pregnancies, in turn, yielded an estimated 844 children at age two with no observed physical defect requiring special care and with IQ's of at least 85.[3*]

A follow-up of a sample of these same children eight years later has now been completed. The two-year assessments of congenital physical defects and mental retardation were confirmed for the most part. Although some original handicaps had yielded to treatment by age ten, enough additional defects had become manifest or acquired to reduce the number without such problems from 844 per 1,000 liveborn at age two to 832 at age ten. When children with serious learning handicaps and emotional problems are added, the number of ten-year-olds without serious problems declines from 832 to 660 per 1,000 live births.

The main purposes of this paper are twofold: to report (1) how children with prenatal and natal defects noted by two years of age have fared, and (2) what the prevalence (case load) of various types of handicapping conditions at age ten is for this community's children.

The setting and the characteristics of the population of Kauai have been described in previous papers.[1-3] It is sufficient to point out that the children studied here are of several ethnic origins (Japanese 1/3, Hawaiian and part-Hawaiian 1/4, Filipinos 1/5, and Caucasians about 1/10). The population's health indexes and the availability and quality of medical care and public health services compare favorably with progressive mainland communities. In the course of this follow-up study we found that only seven of a sample of 750 children were in need of medical care, and this was largely because of delay or negligence by parents, or because treatment plans had not been completed. This

* Number differs slightly from that reported previously. Clinically premature not included as handicapped and rates based on survivors to age two.

speaks well for the physicians on Kauai and for the effectiveness of the health department's Crippled Children's program.

This follow-up study, as well as the earlier Kauai Pregnancy Study (KPS), was closely related to all the existing sources of information about the children: their physicians, the hospitals, schools, and health and welfare departments, all of which provided information from their records and, of course, the parents who also had been interviewed a number of times in the pregnancy study. The local study staff was widely known and trusted, and the few refusals were among parents who had moved from Kauai. We agreed that general findings of the study and those of significance concerning individual children would be made available to all those concerned. Reports of special diagnostic examinations went to the child's physician, psychological examinations to the Department of Special Services of the schools, and letters to the parents with suggestions for follow-up where indicated. With parental consent, reports also were sent to the Division of Mental Health and other care-taking agencies.

Study Group

The *time sample* consists of 750 children, nearly 90 per cent of the island's 837 live births occurring in 1955* (Table 1). Prevalence rates are based on this time sample. An additional *selected sample* is made up of 262 "high-risk" children from a later cohort of 1,104 births.* The latter includes all the two-year survivors in this cohort who had substantial birth defects, injuries or handicaps, birth weight under 2,500 gm, IQ scores under 85, and those considered by either the examining pediatrician or psychologist to be below normal mentally. The combined groups are used in comparing the children's status at ages two and ten.

By the time of the follow-up study 140 of the children (14 per cent) had moved from Kauai to one of the other islands of Hawaii. These are included in the study. We did not attempt to locate the 65 who had moved from Hawaii, mostly to the United States mainland.

Data Collected

Use of already existing information made it possible to eliminate a number of routine procedures and to concentrate on gathering new data.

Table 1—Study groups for ten-year follow-up study, Kauai, Hawaii, 1965-1966 (see text for description of samples)

Time Sample—1955 Live Births		
Total live births in cohort	837	
In first two years:		
Died	12	
Confidential adoptions	4	
Moved from Hawaii	20	
Living in Hawaii at age two	801	(95.7%)
Between two and ten years:		
Died	3	
Moved from Hawaii	45	
Parents refused follow-up	3	
In follow-up study at ten years	750	(89.6%)
Selected Sample—From 1956-1957 Live Births		
Living in Hawaii at age two	276	
Between two and ten years:		
Died	2	
Moved from Hawaii	10	
Parents refused follow-up	2	
In follow-up study at ten years	262	
Combined Samples		
In follow-up study at ten years	1,012*	

* More specifically, the time sample cohort is composed of children born alive to mothers whose last menstrual period (LMP) occurred between the second lunar month of 1954 and the fourth month of 1955. The selected sample was chosen from a cohort of the next later pregnancies: LMP from the fourth lunar month of 1955 to the 12th of 1956.

* Excludes some temporary residents included in previous papers.

The final study design followed the experience gained in a pretest in which all the procedures and protocols were tried out in a group of 63 children just older than the study groups. As a result of this experience, we eliminated the originally planned routine pediatric examination of each child because we discovered nothing pertinent to our study beyond that recorded in the extensive medical and school records already available.

The field staff consisted of a pediatrician, a child psychologist, a public health nurse, part-time interviewers, and clerical personnel. They collected pertinent information for each of the 1,012 children from the following *existing* sources:

1. Records of KPS, the Bureau of Crippled Children, the Divisions of Mental Health and Mental Retardation, the schools' Departments of Special Services, and the Welfare Department.
2. School records including grades, previous intelligence and achievement tests, results of hearing, speech, and vision tests and of physical examinations, and behavior observations.
3. Records of physicians and hospitals for children who had had illnesses or accidents.

The staff obtained *new* information for each child from:

1. A questionnaire filled in by the current teacher (or home room teacher if more than one) including grades in reading, arithmetic, and writing (grammar and spelling), and a check list of physical, cognitive, and emotional problems.
2. A home interview with the mother, covering illnesses, accidents, and hospitalizations in the interval between two and ten years including the names of attending physicians and hospitals, changes in the home environment, and the mother's impressions of the child's school performance and any intellectual and behavior problems.
3. Results of two group tests, the SRA Primary Mental Ability test, elementary level (PMA),[4] and the Bender-Gestalt test.[5]

The professional staff reviewed each child's record to determine completeness of information, evidence of any physical defect or handicapping condition, or any emotional or intellectual problem, and whether they needed additional information or further diagnostic procedures to complete their appraisal of the child's present status. They judged that 299 children (30 per cent) needed diagnostic examinations. Each was carried out by an appropriate specialist (psychologist, pediatrician, audiologist, ophthalmologist, neurologist, and the like). The largest number, 62 per cent, were special psychological examinations; 20 per cent pediatric; 7 per cent speech, hearing, and vision; and 11 per cent other types.

After receiving the results of these special examinations, the panel again reviewed the records. They prepared a final description of any problem, assessed each child's status (physical, intellectual, emotional, and school achievement), and the effect of any existing handicap on school progress, and estimated the need for various types of future care.

The staff examined older children first, in so far as possible, to equalize age at time of screening examinations. When the PMA test was given, two-thirds of the children were ten years of age and over 80 per cent were in the fourth or fifth grade. Of those in the time sample, 733 were in regular school and the remainder, 17, either in Hawaii's institution for the mentally retarded, or attending special schools or classes for the handicapped, or being cared for at home.

Findings

I. Ten-Year Status of Children with Congenital Defects at Age Two

In the present study we compared each child's two-year medical diagnoses, IQ scores, and severity of handicap with similar data obtained at age ten. IQ scores at two came from the Cattell Infant Intelligence Scale.[6] Low PMA IQ scores at ten were checked by the Wechsler Intelligence Scale for Children (WISC).[7]

At age two, we had classified 81 children as having significant congenital defects: 38 severely handicapped and 43 moderately handicapped. Their status at age ten is discussed below.

1. Children Severely Handicapped at Age Two

This group of 38 children consisted of (a) 21 with serious physical-sensory defects without evidence of mental retardation (IQ at least 70), (b) eight with physical defects and mental retardation, and (c) nine who were mentally retarded without serious physical defects.

(a) Physical-Sensory Defects with IQ 70 or Above—Defects included were spina bifida, atresias of the gastrointestinal, genitourinary, and auditory systems, congenital heart, congenital deafness, cleft lip and palate, and cerebral palsy.

Of the original group of 21, 17 diagnoses were confirmed. At age ten, four-fifths of the confirmed cases were still severely handicapped. All had received extensive specialist medical care, mostly under the auspices of the Crippled Children's Division, and required continuing special care. Some surgery was still in progress. Not surprisingly, by age ten half of this group had developed additional defects not manifest or not detected at age two (convulsive disorder, hearing, and vision defects). One child with Sturge-Weber syndrome, with an IQ of 55, was attending a special class for the mentally retarded. The other 20 were in regular school but only half were progressing satisfactorily.

(b) Physical-Sensory Defects with IQ Below 70 at Age Two—These eight children were multiply handicapped with developmental defects of the central nervous system, congenital endocrinopathies, or congenital deafness and blindness, and were severely retarded as well. At two their IQ scores ranged from 16 to 61 (median under 20).

By ten years of age, all previous diagnoses were confirmed. All these children had received extensive diagnostic and treatment services, mostly under the Crippled Children's program, and all still required care. Half of these children had additional physical defects not found at age two (severe visual and hearing problems and epilepsy). All but one had an IQ below 60 (median under 40) and were in special schools or in the institution for the mentally retarded.

(c) Mental Retardation Without Serious Physical Defects at Age Two—At age two, the median IQ score for these nine children had been 64 with a range of 47-69. By age ten, the IQ's of all these children but one had improved some, but only one child had an IQ over 85 (median 77 with a range of 41-98). All were academically retarded, and the two with IQ's in the 40's were in a training school for the mentally retarded. Only one had developed a physical defect, a hearing loss from otitis media.

The above comparisons reveal differences between those whose mental retardation was accompanied by organic defects and those without such defects. As pointed out by others,[8] IQ scores of the former were lower at age two and remained low at ten, while those without physical defects were of a more moderate degree of retardation and their IQ scores tended to improve more by age ten.

2. Children with Less Severe Defects at Age Two

Half of this group of 43 had been children with suspected strabismus. The remainder had consisted of cases of hernia, hydrocele, thyroglossal duct, pilonidal sinus or orthopedic defect.

By age ten, for less than half of the previously suspected strabismus cases, had any eye problem been diagnosed. The overdiagnosis may have been due, in part, to the difficulty of detecting strabismus in Oriental children. All had received diagnostic examinations and treatment services when recommended

(corrective lenses, surgery). Most of the confirmed early strabismus children were still visually handicapped despite treatment. In a few instances treatment had been delayed so that good results were unobtainable.

All those with other types of defects had received recommended care, principally surgery, and none had other than a mild residual defect.

School achievement for these children differed little from achievement of the total study group.

II. Additional Handicaps at Age Ten

In reviewing the status of ten-year-olds in the time sample of 750 who at age two were defect-free, we found 18 children with congenital defects or retardation not noted by two, and 16 with acquired defects.

1. Children with Congenital Handicaps or Mental Retardation Not Evident at Age Two

Of these 18 children, one had mild spastic quadriplegia diagnosed at six years, and one had multiple endocrinopathy diagnosed at eight years. In addition, the IQ's of four, previously scoring from 71-96, had dropped to the 60's. The panel judged all of these children to be severely handicapped requiring special medical or educational services. An additional 12 children were moderately handicapped. Nine of these had eye problems, and failure of early diagnosis and treatment probably resulted in more handicapping than necessary. Two others had developed epilepsy and one an orthopedic defect. All will continue to need specialist care periodically.

(In addition to the above, a minor cleft palate affecting speech, a congenital hip anomaly, and an undescended testicle were diagnosed after age two but had been corrected prior to follow-up.)

2. Children with Acquired Defects

Sixteen children without abnormal findings at age two had acquired handicaps due to accidents (blindness of one eye, organic epilepsy, orthopedic defect), infections (hearing loss due to chronic otitis media, uveitis, chronic urinary tract infections, tuberculosis), and other causes (Legg-Perthe's disease, Letter-Siewe syndrome, hemolytic anemia, severe asthma). Urological investigation failed to reveal congenital anomalies in the three children with chronic urinary tract disease. The panel considered that only the three with organic epilepsy, hemolytic anemia, and Letter-Siewe's disease were severely handicapped. None of the 16 had an IQ score under 70 and only one a score under 85.

III. Prevalence Rates at Age Ten

Of the 750 children in the time sample for whom any defect or health problem was noted, those with problems so minor they interfered little if at all with normal functioning amounted to 73 per cent, and most of these were allergies known to be prevalent in Hawaii. In this paper we are concerned with the children with significant problems: the moderately and the severely handicapped.

Some 6.6 per cent of the 750 ten-year-old children in the time sample were moderately or severely handicapped as a result of physical defects or mental retardation (IQ under 70) or both (Table 2). This rate is slightly higher than the over-all rate at age two (5.6 per cent). Some two-year diagnoses were not confirmed, others changed as a result of treatment, and some of the low IQ's had risen. These decreases were more than equalized by additional children whose handicaps had become apparent in the interim, or who had acquired handicaps as a result of accidents or disease.

The comparatively minor role of post-

Table 2—Proportion of children in time sample with moderately or severely handicapping conditions, ten-year follow-up study, Kauai, Hawaii, 1965-1966

	% of children
Children with congenital defects and/or IQ under 70	4.5
Physical defects only	3.0
Physical defects and IQ under 70	0.8
IQ under 70 only	0.7
Additional children with acquired physical defects (none with IQ under 70)	2.1
Total	6.6
Additional children with IQ's 70-84 (none with significant physical defect)	8.5
Total	15.1

natally acquired physical defects, even at age ten, is shown by the fact that they were only half as frequent as handicaps of congenital origin and mental retardation (2.1 per cent vs. 4.5 per cent).

When children with borderline IQ scores (70-84) are included, the per cent handicapped at ten increases from 6.6 per cent to 15.1 per cent. Since these children were academically retarded, 15 per cent is a more realistic representation of the proportion of children handicapped in the school environment.

Hearing Loss

At age ten, eight of the 750 children (1.1 per cent) had significant loss of hearing—more than 30 decibels in one or both ears at any two frequencies between 250 and 2,000, or more than 35 decibels in one frequency. In half, loss was of congenital origin and associated with multiple physical defects. For the remainder, the loss resulted from otitis media.

Vision Problems

The panel judged that 12 of the 750 children (1.6 per cent) had a vision problem adversely affecting their school progress, i.e., uncorrected muscle imbalance or a refractive error 20/40 or more after correction. Blindness from an accident accounted for one, strabismus for two, and myopia or severe astigmatism for five (including one case of amblyopia). In four other children the defective vision was associated with severe multiple defects; one with Marfan's was legally blind.

IV. Nonmedical Services Needed

This paper is concerned primarily with physical health problems, congenital and acquired, and mental retardation of Kauai's fourth and fifth grade students. However, our interdisciplinary panel estimated each child's needs, not only for special medical services but for educational and mental health services as well. This aspect of the follow-up study will be reported in detail in other papers but a few highlights are given here to help place physical health problems in relation to the educational and emotional problems of school-age children.

Problems requiring special educational services, especially reading, are prevalent throughout the United States, and Hawaii is no exception. While the panel estimated that only about 7 per cent of the ten-year-olds in the time sample would continue to need special medical care, they agreed that the need for special educational help was over five times as great—39 per cent. By far the greatest need was for remedial help in basic skills, e.g., reading, arithmetic, grammar, and spelling—32 per cent; a smaller group (7 per cent) required placement in special classes for those with serious learning disabilities or for the mentally handicapped. Fortunately, of the 32 per cent, over half (18 per cent) needed only short-term

assistance which might be provided, at least in part, by volunteer tutors. This leaves 21 per cent with the most serious problems who were in need of long-term, special educational services. Also we found that twice the number of children had serious emotional problems interfering with school progress—13 per cent—than the number needing special medical care. With the overlap in these two types of needs, we estimated that almost one-third of all Kauai's ten-year-olds needed long-term educational services, or mental health services, or both.

Discussion

We were impressed by the great amount of useful information in existing records of practicing physicians, hospitals, health and social agencies, and schools. Researchers commonly ignore such sources and expend great amounts of time and money repeating examinations and devising elaborate methods for obtaining data that are already recorded. A point of special interest was our finding in the pilot study that we could safely eliminate routine pediatric examinations on all the children by making full use of the records of the physicians, the schools, and the health department. It was far less costly and more efficient to establish good relationships with existing sources of information than it would have been to repeat every procedure. This enabled us to concentrate on collecting new data.

A point needing emphasis is the "high batting average" of the pediatrician-psychologist screening procedures at age two in identifying serious congenital defects and mental retardation. The point to be emphasized is the fact that the procedures used were screening procedures, not thorough diagnostic examinations which would not be feasible for large-scale application. The principal weakness was in connection with eye problems. In view of the importance of early detection of children with serious vision problems, in these early examinations greater attention should have been placed on special sensory screening procedures.

The findings of this follow-up study should encourage those who believe that a great contribution to child health could be made by providing every child with at least one complete physical-sensory and psychological screening examination during the early years.

However, even given early diagnosis and prompt treatment of significant congenital defects, we must not fail to recognize that these handicaps are responsible for only a small proportion of problems interfering with school progress. As suggested by others,[9] handicapping emotional problems requiring treatment were almost twice as prevalent as medical problems, and education problems were over five times as prevalent.

In terms, then, of the needs of children of school age, it is clear that much greater emphasis must be placed on early identification and alleviation of deficiencies in the environment that produce educational and emotional difficulties.[10] These areas require as much if not more attention than has in the past been given to physical health.

Summary

This is a report of a ten-year follow-up of 1,012 children previously studied in the Kauai Pregnancy Study from early in mothers' pregnancies through a two-year pediatric and psychological assessment of physical and mental handicaps of congenital origin.

The earlier diagnoses and predictions for the severely defective children, physical or mental or both, were largely confirmed at age ten. All had received extensive diagnostic and treatment services and three-fourths required special education or institutional care.

IQ scores of the mentally retarded children with organic defects were very low at two years (median under 20),

while those without evidence of organic involvement were higher (median 64). At age ten, the former were still low (median under 40) while the latter had improved somewhat (median 77).

The poorest predictions involved children with eye problems. For only half of those believed to have strabismus by age two had any eye problem been diagnosed by age ten, and an equal number of additional eye problems had been diagnosed by that time. Among the eye cases were children whose vision problems were severe enough to affect school progress. Some of these might have been prevented by earlier diagnosis and treatment.

Pediatric and psychological screening in early childhood, if augmented by careful vision and hearing screening, will identify most children with congenital defects and retardation who need special medical and educational services.

In a time sample of 750 children ten years of age, 6.6 per cent were handicapped significantly as a result of physical-sensory defects or mental retardation (IQ under 70) or both. An additional 8.5 per cent had borderline IQ scores of 70-84 and were academically retarded.

Acquired physical handicaps resulting from accidents or disease were only half as frequent as handicaps of congenital origin and mental retardation, 2.1 per cent vs. 4.5 per cent.

Hearing and vision problems significantly interfering with school performance amounted to 1.1 per cent and 1.6 per cent, respectively.

Over five times as many children required special educational services (39 per cent) as those who required special medical care, and almost twice as many had serious emotional problems interfering with school progress (13 per cent). The greatest need of these ten-year-olds—almost one-third of them—was for long-term educational or mental health services or both. It is clear that much greater emphasis must be placed on early identification and alleviation of deficiencies in children's environment that produce educational and emotional disabilities. These needs require as much if not more attention than has in the past been given to physical health.

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